Case Report

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RECURRENT TRAUMATIC BONE CYST – A CASE REPORT

Swaminathan C*[1]., Mathew Jose[2]., Dhineksh Kumar[3] and Shameem Jamal[4]

[1]Department of Oral and Maxillofacial Surgery, JKK Nattraja Dental College, Komarapalayam
[2,3]Department of Maxillofacial Surgery, Sree Mookambika Institute of Dental Science, Kulasekaram
[4]Consultant Oral and Maxillofacial Surgeon, Trivandrum

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ABSTRACT

The traumatic bone cyst is a rare lesion occurring in the maxillofacial and apendicular skeleton. In maxillofacial skeleton the commonest bone involved is mandible. Occurrence of Multiple traumatic bone cyst is rare and the recurrence of traumatic bone cyst is a rarer phenomenon. Here we present a case of report of recurrent traumatic bone cyst which showed series of recurrence after primary exploratory surgery with reviews of literature.

INTRODUCTION

The term unicarmal bone cyst meaning single chamber bone cyst was introduced in 1876.[1] The traumatic bone cyst was first jntroduced by Lucas and Blum in 1929.[2] Later as the research regarding their causative factor grew their terminologies grew in number as solitary bone cyst, hemorrhagic bone cyst, unicarmal bone cyst, extravasation bone cyst, progressive bone cyst.[3] In 1971 WHO accepted the term Simple bone cyst, later 1992 the term solitary bone cyst was accepted. In 2005 WHO classified solitary bone cyst as bone related lesion with cherubism, central giant cell granuloma, hyperparathyroidism, fibrous dyslplasia.[4] Here we discuss a case of recurrent solitary bone cyst with pertinent review of literature.

Case report

A 50 year old female walked into department of Oral an maxillofacial surgery of Sree Mookambiga Institute of Dental Science, with complaints of pain in right lower back tooth region of jaw for past one week without a significant medical history. On examination the patient had missing teeth 36,46,15,16 and swelling in relation to 34 - 36 and 44 – 46. It was both buccally and lingual expansile lexion of 3x3x2 cm in size, the surface of swelling was normal, on palpation it was tender and no warmth and hard in consistency. The aspirate revealed a blood tinged straw coloured fluid with protein content of 5.4gm/dl. Ortho pantanogram showed well defined radiolucencies in relation to 34 -3 6, 44 – 46, 11-12, 21-22 and 47-48.

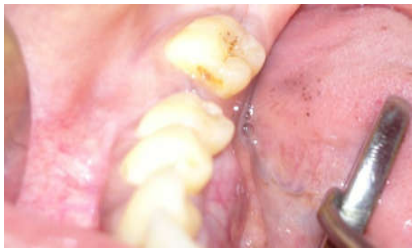

Fig 1 intra oral view of 44-446 region

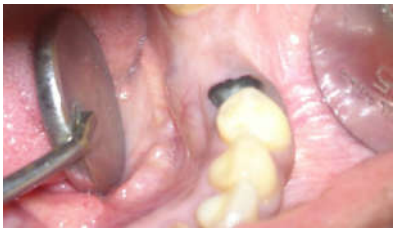

Fig 2 Intraoral View of 34-36 region

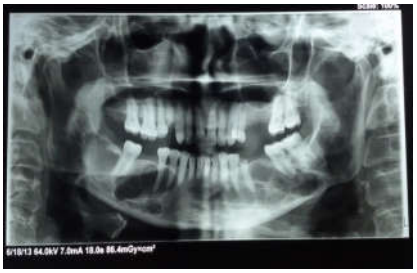

Fig 3 Pre - op Orhtopantanogram

Corresponding author:* **Swaminathan C
Department of Oral and Maxillofacial Surgery, JKK Nattraja Dental College, Komarapalayam

The biochemical investigations like serum calcium and alkaline phosphatase level was normal. A biopsy was done in region of 44-46 to find no epithelium in the cystic cavity.

Fig.4 Straw coloured aspirate

Fig.5 cystic cavity on exploration in relation to 34-36 region.

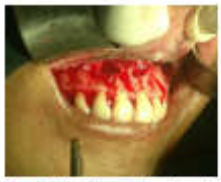

Fig.6 Cystic cavity in relation to 11-12,21-22 region

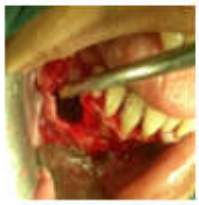

Fig.7 Cystic cavity in relation to 44,46

Fig.8 Cystic cavity in relation 47-48 region

Thus surgical exploration curettage was done in all cystsunder general anaesthesia and the contents from the cyst were found to be connective tissue matrix without epithelium. Thus the diagnosis was multiple solitary bone cyst was established.

The patient was under regular follow up once in a month for the first post operative year. After six months OPG showed considerable bone fill in all the regions and a moderate bone fill in 44-46 region. One year later patient experienced pain in 44 -46 region and patient's OPG showed a radioluceny in same 44-46 region and surgical reexploration was done under local anaesthesia which showed no cystic lining.After two years patient had complaints of pain in right lower back tooth region of jaw and on intraoral examination showed a swelling of size 0.5x0.5x1cm in relation to 44-46 muccobuccal fold which was tender andhard in consistency and aspiration was negative. The OPG showed multilocular radiolucency in relation to 44-46 region of size 1x0.5cm. biochemical insvestigation like serum calcium, phosphorus and alkaline phosphatise was normal. Minimally invasive fenestration procedure was done in region of 44-46 under local anaesthesia which relieved patient pain postoperatively.

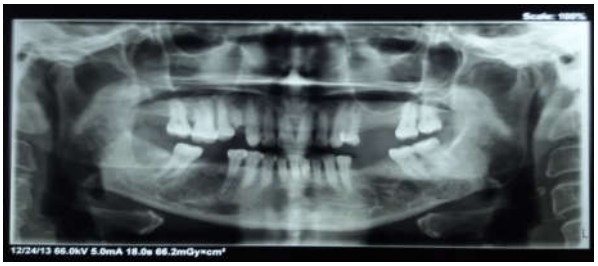

Fig 9 6 months post op

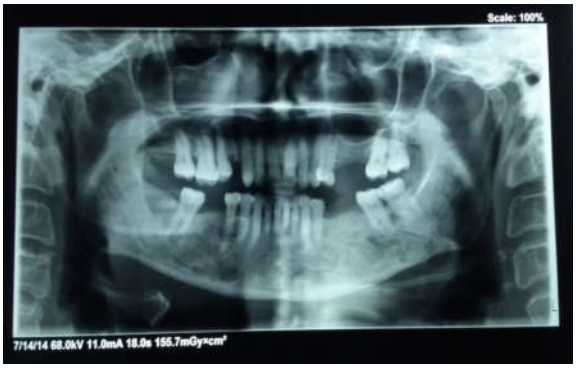

Fig 10 One year post op – cyst in 44-46 region

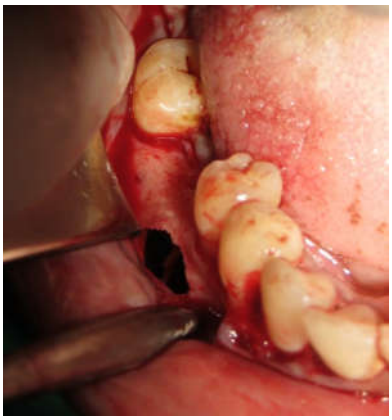

Fig 11 Reexploration after one year

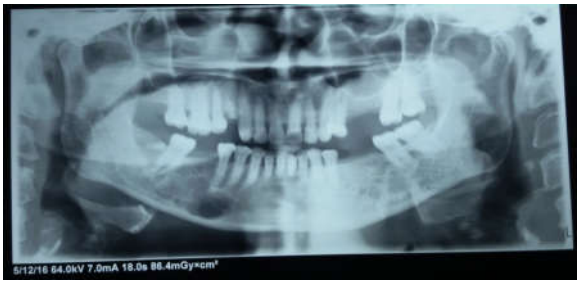

Fig 12 two year post op periapical region of 44 -46

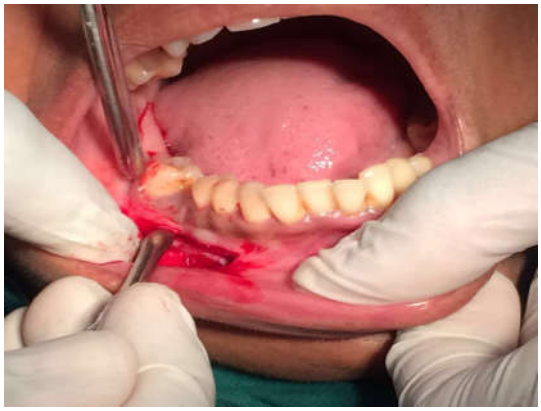

Figure 13 fenestration prepared in the relation to 46

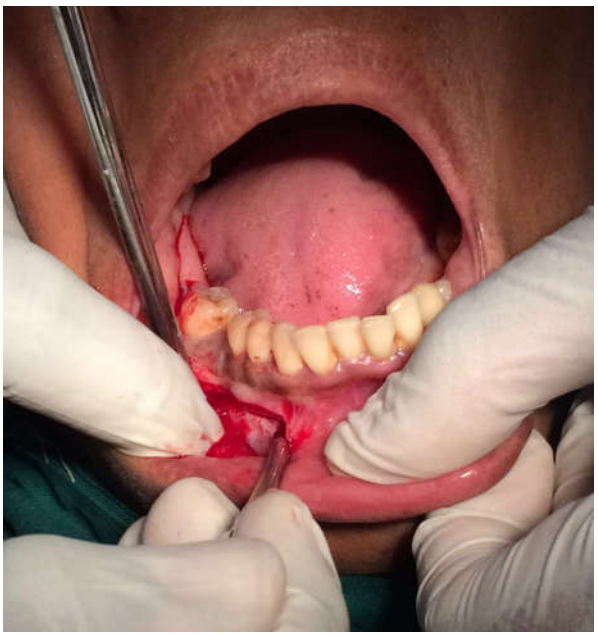

Figure 14 fenestration prepared in relation to 44

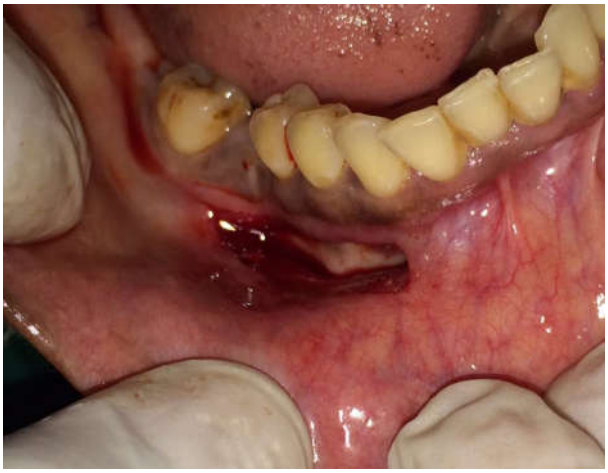

Figure 15 fresh bleeding in the cyst induced

DISCUSSION

According to Ahmed KA, Al-Ashgar F[5], Causative factors for solitary bone cyst were

- Trauma
- Chronic infection
- Fatty marrow necrosis
- Deranged calcium metabolism
- Degeneration of bony lesion
- Developmental anamoly
- Cohen *et al*[6]. described Lack of venous drainage as a cause for TBC.
- Mirra *et al*[7]. described Synovial fluid entrapment as a cause for TBC..

Among the above trauma is the most accepted etiologic factor explained by Ohle *et al*.[8], as minor insignificant trauma causes haemorrhage from marrow, later becoming a clot when the clot increase in size, affects the blood supply to marrow thus causing marrow necrosis. The clot remain unorganised due to absence of nearby healthy connective tissue, it stimulates osteoclastic activity to resorb the bone around to reach the periosteum. Thus cortical perforation occurs and clot organises to healthy mesenchymal granulation but if the clot liquefies before the cortical perforation an empty cavity remain with the cortical borders. Heimdhal [11] noted the time interval between trauma and cyst formation varies from 1week to 20 years.

The mean age of occurrence is 24.3 years with no sex predilection and common sites are poaterior mandible and symphysis and in one fifth of the patient it occurred bilaterally and 11% of them showed multiple cyst. According to Kraugar G E and Cale A E .[9] These are often asymptomatic without root resorbtion and tooth displacement and revealed in radiographic examination appearing as unilocular or multilocular well defined radiolucency. Matsumara *et al*.[10], classified the solitary bone cyst in to two type type A and type B based on etiology. Type A caused by medullary hemorrhage and type B from pre-existing bone lesion like osteogenesis imperfecta. Late Furkaxa *et al*.[10], introduced type C which was froma pre existing fibrous dysplasiaType B and C showed higher recurrence than Type A.

The cystic content may vary from gaseous, seorsanginous fleid and also connective tissue[8]. According to Kuhmichel A, Bouloux GF[12] the contents of the cyst varry based on the duration of existence of the cyst, during early period it is filled with serosanginous fluids and later the quantity of fluid reduces in size and replaced by air and gaseous content. According to Toller *et. al*.[13], the bony walls are permeable to fluids as the intracystic osmotic pressure due to hemorrhage is higher then extracystic osmotic pressure thus attracting more fluid. Radiologically traumatic bone cyst produces a festooning or scalloping effect even in edentulous regions.

The diagnostic criteria according to Rushtom M A et al.[14], 1946

- Lack of cystic lining in surgical exploration
- Unilocular with No content
- Surrounded entirely by bony walls Hansen *et al*.[15], in 1974
- Unilocular or multilocular
- Empty cavity
- May contain soft tissue in addition to transudate

The non surgical management involves allowing spontaneous resolution[16], this applies to the cyst with cortical perforation and steroidal injection like methyl predisolone showing 5% recurrence as advocated by orthopedic literature when compared to surgical enucleation showing 35% recurrence[12].

The surgical treatment involves exploration, curettage, fenestration, cryosurgery and bone grafting the defect with autogenous, allogenous or alloplasticmaterials[12,17]. Surgical exploration treats the lesion by inducing rebleeding and draping the clot with adjacent healthy periosteum. The fenestration is a process of creating bony widows to make the clot to come in contact with the healthy periosteum. Curettage is done only to confirm the absence of any epithelium or in suspected granulomatous change and has a disadvantage of devitalizing the adjacent teeth. Grafting the cystic cavity aids in implant placement but being radio opaque has the disadvantage of interfering in follow up radiograph for recurrence, there by not favouring the prognosis[19].

Multiple traumatic bone cyst was reported in literature by kauger *et al*. With prevalence of 11% and Levin *et al*., reported a case of multiple traumatic bone cyst in association with osteogenesis imperfecta[21] while Anne Cale Jones, Ronald A Baughman and Vijayakumar *et al*., reported a case of multiple traumatic bone cyst with amelogenesis imperfecta.

The recurrence of solitary bone cyst was according to kuroi *et al*.[18], was 4 out of 225 and Seu *et al*.[17], reported recurrence rate of 20% especially when the cyst showed multilocularity, cortical expansion and loss of lamina dura. Horner *et al*., described that reason for recurrence was that the remaining bony cause after surgery and Linday *et al*., reported the presence of nonvital tooth associated with recurrence[18]. Chiba *et al*., in 2002 reported a case of traumatic bone cyst conversion to a central giant cell granuloma.[20] A combination of graft materials like gelfoam, hydroxyapatite and autologous blood mixture was found effective than single material especially for the recurrent lesions.

Review for the solitary bone cyst patients according to Sapone and Hansenwas in first year once in four months and in second year once in six months and in third year once in a year. Later it was modified by Suei *et al*., as for asymptomatic patient the first radiographic evaluation after 12- 17 months and later once a year.

CONCLUSION

Thus multiple solitary bone cyst are rare condition affecting maxillofacial skeleton and diagnosed at the time of surgery. Thus traumatic bone cyst should be considered as differential for any radiolucent lesion occurring in the jaws both in symptomatic and asymptomatic cases.

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